

## On Oral Health in Children and Adults with Myotonic Dystrophy

### AKADEMISK AVHANDLING

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The thesis is based on the following papers:

- I. **Engvall M, Kiliaridis S, Mejersjö C.** Dental needs of patients with myotonic dystrophy. *Swed Dent J.* 1991;15:171-8.
- II. **Engvall M, Birkhed D.** Oral sugar clearance and other caries-related factors in patients with myotonic dystrophy. *Acta Odontol Scand.* 1997;55:111-5.
- III. **Engvall M, Sjögreen L, Kjellberg H, Robertson A, Sundell S, Kiliaridis S.** Oral health in children and adolescents with myotonic dystrophy. *Eur J Oral Sci.* 2007;115:192-7.
- IV. **Engvall M, Sjögreen L, Kjellberg H, Robertson A, Sundell S, Kiliaridis S.** Oral health status in a group of children and adolescents with myotonic dystrophy type 1 over a 4-year period. *Int J Paediatr Dent.* 2009;19:412-22.

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# On Oral Health in Children and Adults with Myotonic Dystrophy

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## Abstract

*Background:* Myotonic Dystrophy type 1 (DM1) is a hereditary neuromuscular multisystem disease with varying clinical expressions and severity. The prevalence worldwide is 5-20/100 000. It is characterized by progressive muscular waste and myotonia. Facial weakness is one of the earliest and most constant features. Muscular weakness has been shown to have an impact on oral health in various ways. The molecular basis for DM1 is an unstable trinucleotide (CTG) expansion on chromosome 19. The severity of the disease and time of onset is roughly correlated to the length of the CTG expansion.

*Aim:* The overall aim of this thesis is to shed light on oral health with focus on periodontal disease and caries in adults and children with DM1. Specific aims are: 1) To assess oral health and motoric ability in adults with DM. 2) To explore caries related factors including oral sugar clearance. 3) To assess oral health and dental care in children with DM1 and to evaluate the changes observed longitudinally over a four year period.

*Subjects and Methods:* In all, 27 adults, ages 35-64 years and 56 children, ages 2.7-18 years, and age and gender matched control persons were examined. Thirty-six of the children with DM and 33 of the control children were examined on two occasions about four years apart. Plaque, gingivitis caries and number of teeth were recorded. In the adult patients, finger force, oral muscular coordination ability, salivary secretion rate and oral sugar clearance were determined. In children, the ability to cooperate during dental treatment was estimated. Questionnaires concerning eating habits and dental care were also used.

*Result:* The adult and children DM1 patients had significantly more caries, plaque and gingivitis and had lost more permanent teeth than the control patients. This was particularly evident for the boys with DM1. Motoric ability, salivary secretion and oral sugar clearance showed less favorable mean values for the adult DM group than for the control group. A negative correlation was found between plaque index and finger force. The children with DM1 had a lower ability to cooperate than the controls and general sedation was often needed during dental treatment.

*Conclusions:* Adults and children with DM1 have more plaque, gingivitis and caries and have lost more teeth than age and gender-matched control persons. This may be explained by lower motoric ability, lower salivary secretion and slower oral sugar clearance and, in children, more cooperation problems. The differences between the groups remained or increased for children with DM1 over the four years in the longitudinal study. For these reasons, intensified prophylactic care, including easy home-care methods, is essential for patients with DM1 to firstly improve their oral health and secondly accustom DM1 children to the dental clinic and the treatment there.

*Key words:* Myotonic dystrophy, dental caries, gingivitis, dental care, oral sugar clearance, behavior management problems.

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